Case Report / Olgu Sunumu

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A rare breast tumor; Adenomyoepithelioma: a case report and review of the literature

Memenin nadir bir tümörü; Adenomiyoepitelyoma: olgu sunumu ve literatürün gözden geçirilmesi

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ABSTRACT

Adenomyoepithelioma is rare benign breast neoplasia characterized by the proliferation of both epithelial and myoepithelial cells of the mammary lobules and ducts. This tumour, which does not have specific risk factors and radiological findings, is mostly seen in advanced ages. This tumour, which occurs with the biphasic proliferation of epithelial and myoepithelial cells, also contains normal breast lobules and ducts. This tumour is very difficult to diagnose and includes many radiological and pathological pitfalls. Although malignant degeneration has been reported in the literature, it is a rare condition. In this study, we present a rare case with radiologically suspicious findings and pathologically reported as adenomyoepithelioma.

Keywords: Adenomyoepithelioma, breast, stromal tumour

ÖZ

Adenomiyoepitelyoma, meme lobül ve kanallarının hem epitel hem de miyoepitelyal hücrelerin proliferasyonu ile karakterize, nadir görülen benign bir meme neoplazisidir. Kendine özgü risk faktörleri ve radyolojik bulguları olmayan bu tümör çoğunlukla ileri yaşlarda görülür. Epitelyal ve miyoepitelyal hücrelerin bifazik proliferasyonu ile meydana gelir ve içerisinde normal meme lobülleri ve duktuslarını da barındırır. Bu tümör tanı açısından oldukça zordur ve radyolojik ve patolojik olarak birçok tuzakları içerir. Malign dejenerasyon literatürde bildirilmekle beraber nadir bir durumdur. Bu çalışmada radyolojik olarak şüpheli bulgular gösteren ve patolojik olarak adenomiyoepitelyoma olarak raporlanan nadir bir olgu sunduk.

Anahtar Kelimeler: Adenomiyoepitelyoma, meme, stromal tümör

INTRODUCTION

Adenomyoepithelioma of the breast (AME) is a very rare benign breast tumour that was first described in 1970. It is generally seen in the fifth and sixth decades (1-3). While AME is frequently encountered in the salivary glands and skin appendages, it is very rare in the breast. Clinically, it presents as a single nodule with a rounded shape, irregular contours, and a hard differential diagnosis with breast cancer (4,5). Histopathologically, AME may exhibit different growth patterns such as tubular, papillary, solid, or more often a combination of these patterns (5). Although benign, local recurrence rates are high in this tumour and wide surgical excision is mandatory for diagnosis and treatment. Various metaplasias and some degree of atypia can be seen in the myoepithelial component of AME (6,7). Malignant transformation may be limited to the epithelial or myoepithelial component, or it can be seen in both components, it has been reported very rarely in the literature and is called malignant AME (7). Here, we present a rare case of AMI presenting with radiologically suspicious findings.

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CASE

In the physical examination of a 55-year-old female patient who applied to the outpatient clinic with the complaint of a right breast mass, a firm mass of approximately 2 cm in size was palpated in the right breast at the 3 o'clock position. In the localization described in the radiological images, a 20x15 mm sized, lobulated contour, hypoechoic, solid mass was observed and was considered suspicious and histopathological verification was recommended. There was no history of breast cancer in the patient's first-degree relatives and there was no other disease in his history. After the tru-cut biopsy result showed sclerosing adenosis, lumpectomy was decided and the mass was excised with wide surgical margins. Histopathological examination revealed that the tumour consisted of small epithelial cells and glands with eosinophilic cytoplasm surrounded by clear cell myoepithelial cells (**Figure**). A diagnosis of adenomyoepithelioma was made in the presence of clinical, radiological and pathological findings. No recurrence or metastasis was observed in the 24-month follow-up of the case.

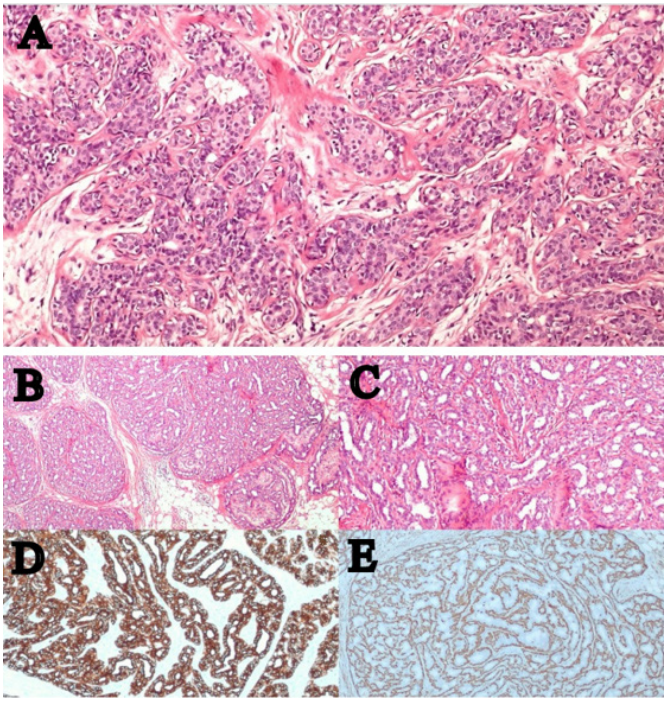

Figure . General view of Adenomyoepithelioma. A-B-C: The tumour consisted of gland structures with eosinophilic cytoplasm surrounded by myoepithelial cells in a loose edematous stroma (x10-x20, H&E). D: Positive staining with CK7 was observed in the epithelial layer of tumoral cells (x10). E: Positive staining with p63 was observed in the myoepithelial layer of tumoral cells (x10).

DISCUSSION

Breast-localized AME was first described by Hamperl in 1970 and is an extremely rare neoplasm, with approximately 150 cases described in the literature (1-3). The age range is wide (26-81), and the incidence increases with age. AME presents as a palpable, well-circumscribed, firm mass (mean 1–2 cm) that can reach up to 8 cm in size (4,5). It is usually localized in the middle of the breast and very rarely presents as satellite nodules, multiple breast masses, or bilateral involvement (6,7). Rarely, the male breast may also be affected (7). In our case, the lesion was presented as a single, well-circumscribed mass of 2 cm in the middle-outer part.

AME is characterized by the proliferation of epithelial and myoepithelial cells of the chest lobules and ducts (8). Myoepithelial cells form part of the normal microscopic anatomy of the breast and are commonly found in the breast. The radiological appearance of this tumour varies considerably and its differential diagnosis from other breast tumours is difficult (8,9). In the US, they are usually seen as a well-circumscribed, hypoechoic, solid lesion. On MRI, they are observed as a well-defined, lobulated contoured density. On mammography, it is usually in the form of an oval or round isodense mass with shaded edges, but it may not allow visualization of the tumour in dense breast tissue (9,10). Microcalcifications can sometimes be detected in the mass in imaging methods. There are also cases in the literature showing homogeneously increasing masses in patients with benign AMI with a dynamic progressive enhancement curve on MR (10). In our case, the lesion was evaluated as suspicious in imaging methods and a definitive diagnosis was made by histopathological examination.

Fine needle aspiration is often not diagnostic for the diagnosis of AMI. Core needle biopsy is ineffective due to the heterogeneity of AMEs (11). Therefore, the definitive diagnosis is excisional biopsy and histopathological examination of the mass. The histopathology of AME is macroscopical as a well-circumscribed, encapsulated and mobile mass (11,12). Microscopically, it is characterized by the proliferation of epithelial cells and glans with eosinophilic cytoplasm surrounded by myoepithelial cells (12). In epithelial cells, immunoreactions are detected with various keratins, especially CK7 and CK19, and EMA, and in myoepithelial cells with p63, SMA, S100, CK14, and calponin. Estrogen and/or progesterone receptors may also be positive (11-13). In our case, positive staining was observed with CK7 in epithelial cells and with p63 in myoepithelial cells.

AME tumours of the breast are rare tumours with variable behaviour. Benign AME is classified according to its growth pattern as tubular, lobulated, and spindle cell variant (14). Malignant transformation is possible and in this case, the tumour should be treated as breast carcinoma. The radiological findings are not specific and significant on their own and do not allow to distinguish the benign or malignant nature of the lesion (14,15). Morphological and hemodynamic features on MRI provide additional information, but no definitive information. Evidence of malignant development relies on histopathologically

detecting findings such as increased mitotic activity, necrosis, cellular pleomorphism, cytological atypia, and infiltrative margins of the tumour nodules that form the lesion (16,17). Malignant changes may involve mostly epithelial cells and rarely both. The role of immunohistochemistry in the diagnosis of malignant AME is limited (17). Nuclear atypia, nuclear pleomorphism, mitosis, necrosis and infiltration of surrounding tissues were not observed in our case.

Adenosis, intraductal papilloma, nipple adenoma and fibroadenoma should be considered in the differential diagnosis when diagnosing AMI (9). The above-mentioned radiological, morphological and immunohistochemical features of the case are very helpful in avoiding these diagnostic pitfalls (9,10). While malignant cases are frequently described in the literature, benign AMI cases have been reported rarely. Therefore, there are no guidelines to distinguish between benign and malignant cases of AMI, and there is no clear consensus on their treatment (14,15). In our case, typical morphological and immunohistochemical findings were very helpful in the differential diagnosis.

In cases of AMI, the distinction between benign and malignant should be made carefully. Although the prognosis is good for benign AMI, the prognosis is poor in malignant cases due to high recurrence and metastasis (15). Malignant AMEs metastasize to organs such as the lung, brain, and liver by hematogenous rather than lymphatic route. Tubular variants and some lobular variants with high mitosis (>3 mitoses/10 HPF) are associated with a high incidence of recurrence (15,16). Given the uncertain and unpredictable tendency for malignant transformation and the risk of local recurrence, conservative excision with negative margins seems to be the appropriate surgical treatment at present. In the case of malignant AMI, mastectomy and analysis of the sentinel lymph node are recommended. Chemotherapy has not had much success (16,17). No recurrence or metastasis was detected in the 2-year follow-up of our case.

CONCLUSION

AME is a rare breast tumour with a mostly benign prognosis and should be kept in mind in the differential diagnosis with other solid lesions of the breast. Imaging features are not pathognomonic and definitive diagnosis is only possible with histopathological examination. Surgical excision with a solid margin is the optimal therapeutic method because of the high recurrence rate in benign and aggressiveness in malignant ones.

ETHICAL DECLARATIONS

Informed Consent: Written consent was obtained from the patient participating in this study.

Referee Evaluation Process: Externally peer-reviewed.

Conflict of Interest Status: The authors declared that there was no conflict of interest in this study.

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